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ORIGINAL ARTICLE

Prevalence of Dental Anomalies in Deciduous and Permanent Dentition of Cleft Lip and Palate Patients

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ABSTRACT

Objective: To evaluate the association between oral cleft (OC) phenotypes and dental abnormalities (DA) in both permanent and deciduous dentition of patients with Cleft Lip and/or Palate (CL/P). **Material and Methods:** This cross-sectional study included CL/P patients older than 4 years of age who had orthopantomograms good enough for DA evaluation. The DA evaluated were tooth agenesis, giroversion, root dilaceration, conoidism, fusion, ectopic tooth, concrescence, tooth twinning, and dental transposition. Sample size calculation was performed. The prevalence of DA in deciduous and permanent dentition was reported, both for maxillary and mandibular teeth. **Results:** A total of 159 patients were included in this study. There was a statistically significant difference in the prevalence of women and men with cleft palate, being cleft palate more prevalent in women (p=0.005). Regarding sexual dimorphism, no statistically significant difference was found between OC phenotypes (p>0.05). Concerning the prevalence of DA in the cleft region, it was observed that DA are more prevalent in permanent dentition (60%) than in deciduous dentition (24%). **Conclusion:** Oral cleft phenotypes are associated with sex, but dental abnormalities are not, both in permanent and deciduous dentition. The prevalence of dental abnormalities is higher in the cleft region, but mandibular teeth may also be affected. Dental abnormalities in the oral cleft region shows to be more prevalent in permanent dentition than in deciduous dentition.

Keywords: Mouth Abnormalities; Jaw Abnormalities; Tooth Abnormalities.

Introduction

The development of the orofacial region involves a complex series of events coordinated by reciprocal interactions between epithelial and mesenchymal cells [1,2]. Disturbances in signaling pathways may cause oral clefts (OC) [2], a condition that can affect the lips and/or palate in approximately 1 out of 700 live births [3]. OC significantly impact the oral health-related quality of life, especially due to scar formation, dentofacial deformities development, speech impairment, and the higher prevalence of dental abnormalities (DA) [4,5].

Individuals with OC exhibit various DA, such as variation in tooth crown size, tooth agenesis, tooth hypoplasia, and tooth malposition [4-7], affecting both deciduous and permanent dentition [8]. The OC phenotype may affect the prevalence of DA: cleft lip and palate (CLP) has been associated with a higher prevalence of tooth agenesis, cleft lip (CL) has been associated with supernumerary teeth, and cleft palate (CP) with teeth crowding and other dental abnormalities [7]. However, other studies have found that the cleft phenotype does not significantly influence the prevalence of DA [9-11]. Because not only developmental issues may be associated with the frequency of DA, the influence of primary lip surgical procedures in DAs has been studied [10].

Despite the factors involved with DA, patients with OC need multidisciplinary follow-up throughout their lives, including various dental specialties such as pediatric dentistry, orthodontics, periodontics, prosthodontics, oral and maxillofacial surgery [12,13]. One of the reasons why patients with OC need multidisciplinary follow-up is the higher prevalence of DA. Tooth agenesis can complicate orthodontic treatment [12]; conoid tooth may affect not only aesthetics, but also the preparation for a fixed prosthetics; root dilaceration may difficult endodontic procedures. Furthermore, DA, such as structural anomalies, may affect dental hygiene, leading to periodontal disease, tooth loss, and aesthetical and psychological issues [4,13,14]. Due to these complications, a specialized dental team becomes essential.

Systematic reviews have been conducted to investigate the prevalence of DA among patients with OC, and have frequently cited the lack of rigorous methodological aspects and inadequate diagnostic criteria for DA as a limitation [4,15,16]. However, these studies did not evaluate the prevalence of DA in deciduous dentition. To study the correlation between OC and DA in both permanent and deciduous dentition is crucial for a deeper comprehension of the treatment requirements for patients with OC. Therefore, this study aimed to evaluate the association between OC phenotypes and DA both in the permanent and deciduous dentition of males and females with CLP.

Material and Methods

Ethical Considerations

This study was approved by the local ethics committee (Opinion No. 3.441.872) and followed the Declaration of Helsinki for study with humans. All participants or their legal guardians/parents signed an informed consent authorizing their participation. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines was used to report this study.

Study Design and Sample Selection

This cross-sectional study was developed at a reference center for cleft lip and palate care from January 2021 to January 2022. Patients scheduled for ongoing dental treatment during this period were invited to participate. Inclusion criteria were as follows: non-syndromic patients older than 4 years, of any sex, with any

cleft phenotype, and that had digital or analogic orthopantomogram. Exclusion criteria were damaged orthopantomograms that made it impossible to evaluate DA.

Sample size calculation was performed using the website openepi.com version 3 (http://openepi.com/SampleSize/SSPropor.htm) and resulted in a sample size of 159 patients. The population size considered for sample size calculation was 7.200 patients seen in one year, the confidence interval was 95% and the frequency of at least one dental anomaly in the cleft lip and palate population was 12% [17].

Data Collection and Imaging Analysis

Data such as age, sex, cleft phenotype, and type of dental abnormality per tooth were collected. Cleft phenotypes were classified as cleft lip (CL), cleft lip and palate (CLP), and cleft palate (CP). The classification was performed based on clinical and medical records evaluation.

The imaging analysis was performed by two examiners who were calibrated by a senior researcher (GVC). Orthopantomograms evaluations were performed in a high-resolution monitor in a clean room designed to be free from distractions, with magnifying aids available. When analogic orthopantomograms were available, the evaluation was performed in the same room using a negatoscope that provided enough contrast for interpretation. The examiners were submitted to intra- and inter-examiner calibration. The Kappa value for intra-examiner calibration was 0.875 ($p<0.001$) and for the inter-examiner calibration was 0.876 ($p < 0.001$).

The DA evaluated were tooth agenesis, giroversion, root dilaceration, conoidism, fusion, ectopic tooth, concrescence, tooth twinning, and dental transposition. Definitions for dental abnormalities were described by different authors [18-20]. Both deciduous and permanent dentition were analyzed. In instances of uncertainty regarding the identification of a dental abnormality in an orthopantomogram, a senior dental surgeon (RS) was consulted to provide a final diagnosis.

Statistical Analysis

The IBM SPSS - Statistical Package for Social Sciences (IBM Corp., Armonk, NY, USA) Software version 24.0 was used for the statistical analysis. The prevalence of cleft phenotype and the presence of DA (both in deciduous and permanent dentition) were analyzed according to sex – the Chi-Square Test was used. The significance level was set at 95% ($p<0.05$). The analysis of cleft phenotype distribution according to sex was submitted to a Z-test and the p-value was adjusted using the Bonferroni method. To avoid biases, only patients with CLP were included in the DA analysis. If a participant had at least one DA both in deciduous or permanent dentition, he/she was classified as having a DA. The prevalence of dental abnormality in deciduous dentition and the prevalence of dental abnormality in permanent dentition per tooth were reported, both for maxillary and mandibular teeth. Additionally, the prevalence of DA in the cleft region was also reported. The teeth considered to be in the cleft region were central and lateral incisors and canines.

Results

A total of 159 patients were included in this study. Among these, 78 patients had orthopantomograms that permitted the analysis of deciduous dentition (Figure 1). The study included 80 males (50.3%) and 79 females (49.7%) with a median age of 17 years (range: 6 – 64). The most prevalent cleft phenotype in this study was CLP (120 patients, 67.0%), followed by CL (23 patients, 14.5%). This data and the prevalence of every cleft phenotype included in this study is presented in Table 1.

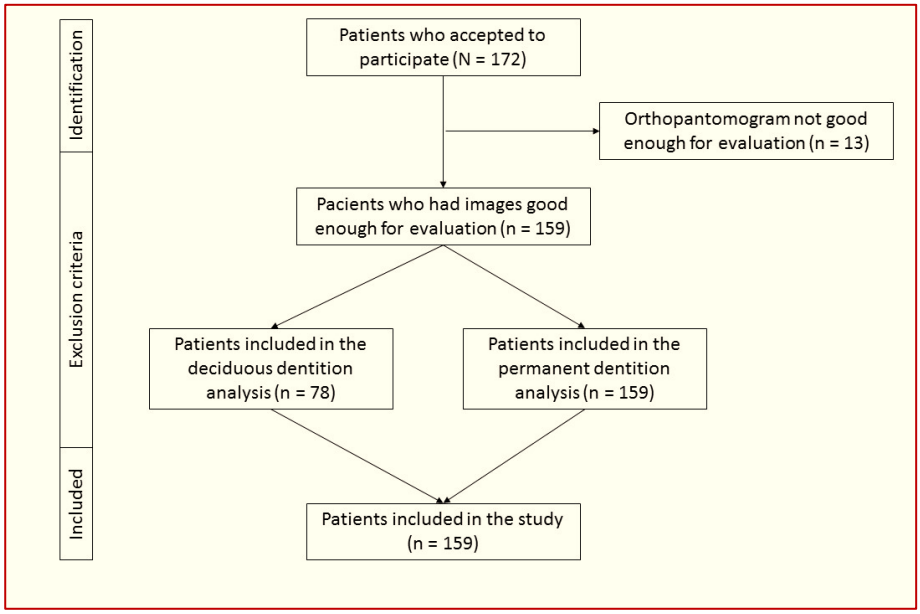


Figure 1. Flowchart showing sample selection according to inclusion and exclusion criteria.

Table 1. Prevalence of cleft phenotype according to sex.

Cleft Phenotype	Male N (%)	Female N (%)	p-value*
CL	14 (17.5) [a]	9 (11.4) [a]	0.005
CLP	64 (80.0) [a]	56 (70.9) [a]	
CP	2 (2.5) [a]	14 (17.7) [b]	
Total	80 (100.0)	79 (100.0)	

CL: Cleft Lip; CLP: Cleft Lip and Palate; CP: Cleft Palate. Different superscript letter denotes a subset of "sex" categories whose column proportion differ from each other; *Chi-square test, p-value adjusted by the Bonferroni method.

The prevalence of at least one DA in deciduous or permanent teeth was 94.2% (n = 120). We found that 25.8% (15 out of 58 patients evaluated for deciduous teeth) presented with at least one DA in their deciduous teeth, while a significantly higher prevalence of 94.2% (113 out of 120 patients) was noted for at least one DA in permanent teeth. No significant association between the prevalence of DA and sex was found (p>0.05), as detailed in Table 2.

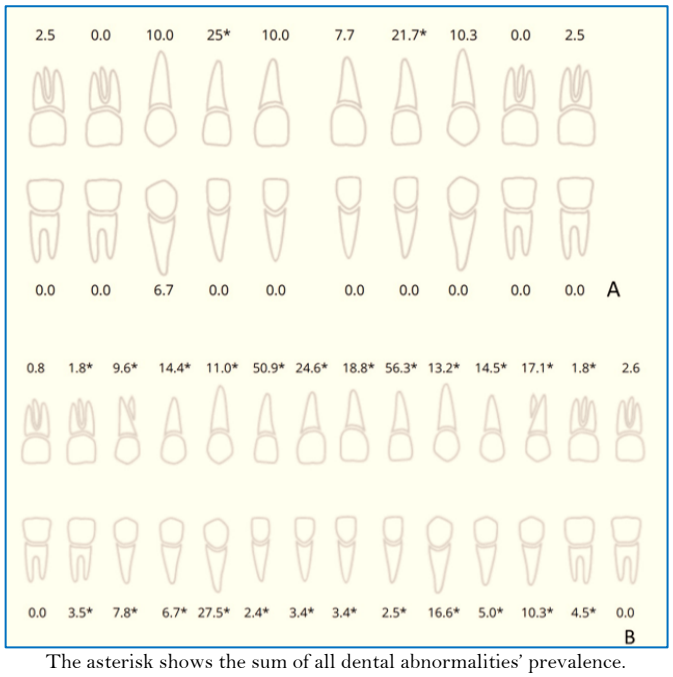
Table 2. Prevalence of dental abnormality in the total population, in deciduous teeth and in permanent teeth of cleft lip and palate patients.

Variables	Male N (%)	Female N (%)	p-value*
Dental abnormality			
Yes	62 (55.0)	51 (45.0)	0.249
No	2 (29.0)	5 (71.0)	
Dental abnormality in deciduous dentition			
Yes	8 (53.0)	7 (47.0)	1.000
No	23 (54.0)	20 (46.0)	
Dental abnormality in permanent dentition			
Yes	62 (55.0)	51 (45.0)	0.249
No	2 (29.0)	5 (71.0)	

*Chi-square test with a confidence level of 95% (p < 0.05).

In the analysis of DA by type, giroversion was the most prevalent (17.8%) in deciduous teeth. The deciduous tooth that presented the highest prevalence of dental abnormalities was the right deciduous maxillary

lateral incisor (17.8% of giroversion, 3.6% of tooth agenesis and 3.6% of root dilaceration), followed by the left deciduous maxillary lateral incisor (13.0% of giroversion, 8.7% of root dilaceration) (Figure 2A). In permanent dentition, tooth agenesis (49.1%) was the most prevalent dental abnormalities. The right permanent maxillary lateral incisor was the most frequently affected by dental abnormalities (49.1%), followed by the left permanent maxillary lateral incisor (40.8%). This data is presented in detail in Figure 2B. The supplementary material shows the prevalence of each dental abnormality per tooth.



The asterisk shows the sum of all dental abnormalities' prevalence.

Figure 2. (A) Odontogram representing deciduous dentition and the prevalence (%) of DA, and (B) Odontogram representing permanent dentition and the prevalence (%) of DA.

When we analyzed the prevalence of tooth abnormalities in the cleft region (central incisor, lateral incisor, and canine), we found a prevalence of 28.0% of at least one tooth abnormality in deciduous dentition and 69.7% in permanent dentition (Table 3).

Table 3. Dental abnormality prevalence in deciduous and permanent dentition in the cleft region.

Dentition	Abnormality in the Cleft Region?	N (%)
Deciduous Dentition	Yes	14 (28.0)
	No	36 (72.0)
Permanent Dentition	Yes	83 (69.7)
	No	36 (30.3)

Discussion

This study was carried out in a state reference center for comprehensive care of patients with cleft lip and/or palate, the second largest center in Brazil. Despite being a primary study and not evaluating the longitudinal relationship between the presence of anomalies in both deciduous and permanent dentition, this study is a pioneer in the investigation of dental anomalies in deciduous and permanent dentition. It was possible to study the prevalence of cleft phenotypes and the prevalence of dental anomalies and to associate them with sexual dimorphism and the region where they occur (whether in the cleft region or not). Another difference in this study was the inclusion of mandibular teeth in the reports of DA data.

The OC occurs due to several factors, including genetic factors [2]. The same genes related to the development of clefts are also associated with the development of teeth [2,21,22]. Our findings reveal a higher prevalence of DA in the mandibular dentition when compared to other studies, both in primary dentition [23] and permanent dentition [24]. Therefore, these descriptive results suggest that there may be an association between the prevalence of dental anomalies in mandibular teeth and the presence of cleft lip and palate. Further studies are suggested.

Regarding the OC phenotype, it was found that the prevalence of cleft palate is more prevalent in females than in males. These results are in accordance with the literature [25,26]. It was possible to observe that although sexual dimorphism is associated with the cleft phenotype, OC is not associated with the prevalence of DA, both in the deciduous dentition and in the permanent dentition. These results are in line with what was found in recent studies [4,11,27], where no association was found between sex and the prevalence of dental anomalies.

Regarding the presence of DA in patients with cleft lip and palate, a high prevalence of dental anomalies was observed in the fissure region of the primary dentition, higher than that presented in studies involving only patients without CLP [23]. As for permanent dentition, it has been observed that it is more common to find dental anomalies than not to find them. This information is important for rehabilitative planning for patients with CLP. The presence of DA, such as the ectopic eruption of teeth in the fissure region, hinders and makes it impossible to perform bone grafting [28]. The non-continuity of the alveolar ridge limits orthodontic movement and prosthetic rehabilitation with osseointegrated implants, leading to the need for rehabilitation with mobile or fixed prostheses, which do not always meet the patient's expectations regarding function and aesthetics [28]. Likewise, the presence of giroversions makes endodontic treatment and prosthetic preparation more difficult, in addition to impairing hygiene. These limitations can reduce the longevity of the proposed treatment, as difficult hygiene can lead to loss of treatment due to caries and periodontal disease.

Among the limitations of the study, we can mention the moment the x-rays were taken. Anomalies would be better assessed if all included patients had an x-ray taken before any dental intervention (whether surgical or orthodontic). This would reduce the possibility of recall bias and improve the assessment of anomalies. Another limitation of the study was the type of imaging exam. Panoramic radiographs present a high degree of distortion, especially in the anterior region of the mandible and maxilla [29]. An alternative would be evaluation using cone beam computed tomography (CBCT) or periapical radiographic survey. As periapical surveys and CBCT are not routinely requested for the treatment of these patients, we would encounter an ethical barrier in recommending these exams solely for research purposes. However, aware of this limitation of panoramic radiography, those patients who had radiographs that did not allow for adequate evaluation were excluded from the study.

Observing the limitations present in the literature, it is suggested that different methodologies be used in future studies, especially considering the moment of radiographic evaluation. To determine the association between anomalies in the primary dentition and anomalies in the permanent dentition, we suggest that longitudinal studies be developed.

Conclusion

Oral cleft phenotypes are associated with sex, while dental abnormalities do not demonstrate a sex association, both in the permanent and deciduous dentition of cleft lip and palate patients. Furthermore, the prevalence of dental abnormalities is higher in the cleft region, but mandibular teeth may also be affected.

Authors' Contributions

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All authors declare that they contributed to critical review of intellectual content and approval of the final version to be published.

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None.

Conflict of Interest

The authors declare no conflicts of interest.

Data Availability

The data used to support the findings of this study can be made available upon request to the corresponding author.

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